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Title

Adenocarcinoma arising at a colostomy site with inguinal lymph node
metastasis: Report of a case

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Running head

Colostomy site cancer with inguinal LN meta

Abstract

Inguinal lymph node (LN) metastasis from adenocarcinoma arising at a colostomy site is extremely rare, and the significance of surgical resection for metastatic inguinal LNs has not been established. An 82-year-old woman who had undergone abdominoperineal resection 27 years earlier was admitted to our hospital complaining of bleeding from a colostomy. Physical examination revealed that a tumor at the colostomy site directly invaded into the peristomal skin, and that a left inguinal LN was firm and swollen. PET/CT scan demonstrated accumulation of ^{18}F -fluorodeoxy glucose into both the colostomy tumor and the left swollen inguinal LN, while there was no evidence of metastasis to liver or lungs. She underwent open left hemicolectomy with wide local resection of the colostomy, and dissection of left inguinal LNs. Histological diagnosis was a moderately differentiated adenocarcinoma that directly invaded into the surrounding skin and metastasized to the left inguinal LN. The patient has been followed up for more than 5 years without any sign of recurrence.

In general, inguinal LN metastasis from colorectal cancers is regarded as a

systemic disease with a poor prognosis, and so systemic chemotherapy and radiotherapy, but not surgical LN dissection, are recommended. Considering the lymphatic drainage route in the present case, inguinal LN metastasis does not represent a systemic disease but rather a sentinel nodal metastasis from adenocarcinoma at a colostomy site. Surgical dissection of metastatic inguinal LNs should be considered to enable a favorable prognosis in the absence of distant metastasis to other organs.

Mini-Abstract

Inguinal lymph node metastasis from adenocarcinoma arising at a colostomy site does not necessarily represent a systemic disease. Surgical dissection should be considered for a favorable prognosis.

Keywords

colostomy site cancer, inguinal lymph node, metastasis, lymph node dissection

INTRODUCTION

Adenocarcinoma arising at a colostomy site is rare with only 10 cases reported in the English literature [1-10]. In addition, isolated inguinal lymph node (LN) metastasis from colon cancer is extremely rare [11,12], and the significance of surgical resection for metastatic inguinal LNs has not been established. Here we reported the first case of adenocarcinoma arising at a colostomy site with left inguinal LN metastasis, which was successfully treated by surgical resection.

CASE REPORT

An 82-year-old Japanese woman was admitted to our hospital in 2009 complaining of bleeding from a colostomy. The patient had hypertension and bronchial asthma that were well controlled by medication, and she had no family history of malignancy. She had undergone abdominoperineal resection of the rectum with a sigmoid colostomy 27 years earlier, and the resected tumor had revealed to be benign by the pathological examination. She had not suffered from any stoma-related complications, such as stomal stenosis or peristomal dermatitis, after the colostomy was formed. A tumor, about 5 cm in diameter, was located at the colostomy site, and directly invaded into the surrounding skin of the colostomy at the left lower abdominal quadrant (Fig. 1). In addition, a firm swelling of the left inguinal LN, about 2 cm in diameter, was observed. Biopsy from the tumor was suggestive of moderately differentiated adenocarcinoma. Laboratory data revealed mild anemia (hemoglobin, 11.2 g/dl) and an elevated carcinoembryonic antigen level (CEA, 23.2 ng/ml; normal level < 5.0 ng/ml). The liver and renal functions were normal. Computed tomography (CT) revealed a

locally advanced colostomy site tumor infiltrating into the surrounding skin with regional LN swelling around the colostomy and an isolated LN swelling at the left inguinal region (Fig. 2a, b). There was no evidence of distant metastasis to liver or lungs. Positron emission tomography/computed tomography (PET/CT) scan demonstrated ^{18}F -fluorodeoxy glucose (FDG) uptake into both the primary tumor at the colostomy site and the swollen left inguinal LN (Fig. 2c, d). In the following month, the patient underwent open left hemicolectomy with wide local resection of the colostomy and dissection of the left inguinal LNs. A new transverse colostomy was constructed in the right lower abdominal quadrant. The resected specimens showed a 5.0 × 3.5 cm Borrmann type III tumor at the distal end of the colostomy, and there was no mucosal abnormality in the rest of the resected colon (Fig. 3). Histopathological examination revealed a moderately differentiated adenocarcinoma infiltrating into the surrounding skin with metastases of a regional LN and a left inguinal LN (Fig. 4a-d). The tumor was classified as Stage IVA (pT4b, pN1a and pM1a) according to the TNM classification of Union for International Cancer Control (UICC) [13]. The patient

had a favorable post-operative course, and was discharged from the hospital without any complication. A month later, the CEA level returned to the normal range. Adjuvant chemotherapy was not performed because of her old age, and she has been well without recurrence for more than 5 years after surgery.

DISCUSSION

Adenocarcinoma arising at a colostomy site is rare and only 10 cases have been reported previously in the English literature [1–10]. Almost all of these cases had a past history of malignancy, mainly in their rectum, and were considered as metachronous multiple colon cancers. The exact etiology for these conditions has not been established because of its rare occurrence, but the following hypotheses have been proposed; 1) adenoma-carcinoma sequence, 2) physical mucosal damage from persistent compression by clothing or stenosis, 3) exposure to enterobacteria and bile acids in stool, 4) cancer family syndrome, 5) *de novo* metaplasia, 6) direct extension of the disease, 7) hematogenous or lymphatic spread, and 8) implantation of exfoliated tumor cells [1,2]. In the present case, adenocarcinoma at a colostomy site was identified as a primary tumor because the rectal tumor resected 27 years earlier had been revealed to be benign. The patient did not have a family history of malignancy, and there was no polypoid lesion or inflammation in the remainder of the resected colon. Histological examination also revealed no adenomatous

components. The function of her colostomy was satisfactory and there was no sign of bowel obstruction. Therefore, the most likely cause of this patient's tumor may be *de novo* metaplasia. We propose that long-term and periodical observation of a colostomy should be performed after construction.

In addition, this patient had inguinal LN metastasis from adenocarcinoma at the colostomy site. Although inguinal LN metastasis from rectal cancer is a rare but well-known clinical occurrence, particularly in cases of anal canal cancer [14,15], inguinal LN metastasis from colon cancer is extremely rare. To date, only two other similar cases were reported; one case arising from cecum cancer, and the other from sigmoid colon cancer [11,12]. An interesting aspect about these particular cases is the lymphatic route by which cancer cells travel to metastasize to the inguinal LN. The usual pattern of regional LN metastasis in colorectal cancer follows the vascular distribution in the mesocolon. In the present case, one possibility is that the tumor invaded into the peristomal skin subsequently metastasizing to the left inguinal LNs through superficial lymphatic pathways along the left inferior epigastric artery. According to the TNM staging

system of UICC, inguinal LN metastasis from colorectal cancer is classified as a distant metastasis (M1, stage IV) and it is regarded as a systemic disease. It was previously reported that inguinal LN metastases from rectal cancer usually occur as a consequence of locally advanced or recurrent pelvic tumors with other distant metastases and have a poor prognosis [14]. Therefore, in general, systemic chemotherapy and radiotherapy, but not surgical LN dissection, are recommended for these patients. In contrast, UICC staging system for the anal canal cancer classifies inguinal LNs as regional LNs because of the direct lymphatic drainage route. It has been reported that the anal canal cancer patients with metastatic inguinal LNs are usually treated with inguinal dissection and postoperative radiation, and that the 5-year overall survival rate of these cases is about 50% [16]. Considering the lymphatic drainage route in the present case, inguinal LN metastasis does not represent a systemic disease but rather a sentinel nodal metastasis from adenocarcinoma at the colostomy site. The effect of local therapy, such as surgical LN dissection and radiotherapy, on the survival of cancer patients has been debated for decades. However, results

of some recent clinical trials support that improved local therapy is causally associated with better overall survival in several types of cancer [17]. In fact, in this present case, long-term survival was achieved without recurrence. Therefore, if there is no metastasis to distant organs other than inguinal LNs, surgical dissection of inguinal LNs should be considered to achieve a better survival.

Conflict of interest statement

None declared.

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Legends of figures

Fig. 1 An ulcerating tumor at a colostomy site invaded into the peristomal skin.

Fig. 2 a, b CT scans showed a tumor invading into the surrounding skin with swollen regional LNs around a colostomy (a), and a swollen left inguinal LN (b).

c, d PET-CT scans showed FDG uptake in both the tumor at a colostomy site (c) and the swollen left inguinal LN (d).

Fig. 3 Macroscopic findings of resected specimen revealed a 5.0 × 3.5 cm Borrmann type III tumor at the distal end of the colostomy, and there was no mucosal abnormality such as adenomatous polyps and inflammation in the rest of the resected colon.

Fig. 4 Histopathological findings of the resected specimen revealed the tumor of a colostomy site as moderately differentiated adenocarcinoma (a: HE ×40, b: HE

×200). The tumor invaded skin (c: HE ×40) and metastasized to a left inguinal

LN (d: HE ×200).

Fig. 1



Fig. 2

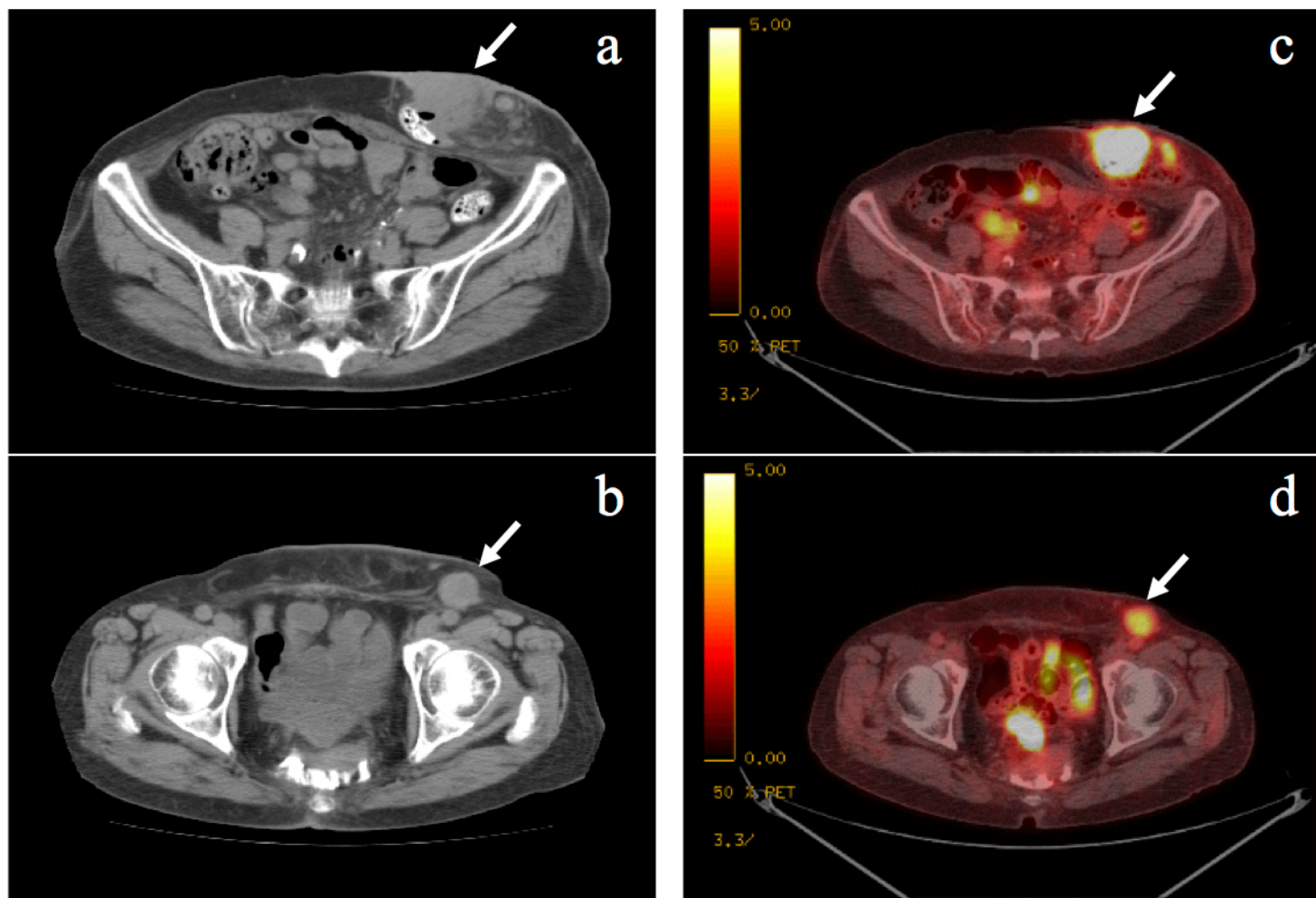


Fig. 3



Fig. 4

